An Uncommon Cause for Common Symptoms of Abdominal Pain and Diarrhea: A Case Report of Human Intestinal Spirochetosis in an Immune-Competent Patient

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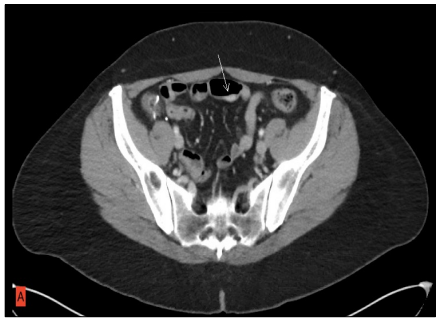
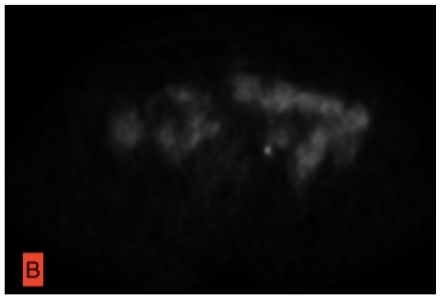
Introduction

Abdominal pain, diarrhea, and weight loss are common but non-specific symptoms that can lead to extensive evaluation for the etiology, e.g., malignancy, infection, or IBS. While the incidence of neuroendocrine tumors (NETs) is increasing, those of the GI tract are rare, and even rarer is human intestinal spirochetosis (HIS). Here, we present an unusual case of an immune-competent patient incorrectly diagnosed with a NET and found to have HIS.

Images





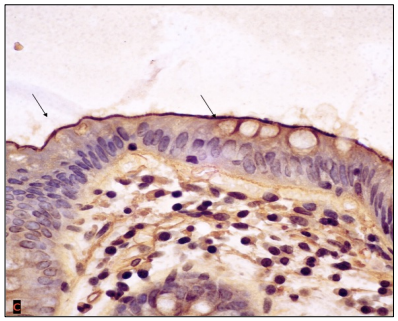


Image A: CT A/P (coronal) showing 1 cm focus of high attenuation/enhancement along the dependent aspect of an ileal bowel loop (white arrow), which may represent hyperdense enteric contents versus an enhancing lesion but is suboptimally evaluated secondary to lack of precontrast images.

Image B: No abnormal Dotatate-avid lesions.

Image C: Focal fuzzy basophilic layer of organisms at luminal surface of colonic epithelium is noted. These organisms are highlighted by Warthin-Starry stain (black band of organisms on the luminal surface of colonic epithelium), as shown with arrow.

Case Description

A 54-year-old female with IBS-D and GERD on PPI was referred for one year of abdominal pain, diarrhea, and weight loss. A year ago, she had a cholecystectomy and made dietary changes without resolution. Prior labs collected showed elevated gastrin and somatostatin but normal VIP and urine 5-HIIA, concerning for NET. Prior double balloon enteroscopy did not locate any bowel lesions. CT A/P showed 1cm focus of high attenuation/enhancement in the ileum (image A). Chromogranin A levels were elevated. PET Dotatate scan performed two weeks later did not show any abnormal lesions (image B) despite persistent symptoms. Surgical oncology recommended repeat gastrin levels off PPI, and she was given colestipol for diarrhea with minor relief. Repeat gastrin levels were normal, and EGD/colonoscopy performed two months later was endoscopically unremarkable with no lesions seen after 30 cm intubation of the TI. However, random colon biopsies showed a focal fuzzy basophilic layer of organisms at luminal surface, highlighted by Warthin-Starry stain, suggestive of intestinal spirochetosis (images C-D). She was prescribed a two-week course of metronidazole with eventual full relief of diarrhea. She was referred to ID for further management.

Discussion

Human intestinal spirochetosis is likely underestimated due to its rarity, especially in heterosexual and immune-competent patients, and suboptimal diagnostic methods. Thus, a subset of IBS patients with diarrhea can often be misdiagnosed for years, especially if they are not immunocompromised. Long-term PPI use is also very prevalent, and one long-standing concern is PPI-induced gastrin elevation secondary to hypoacidity, which confounded our results. Ultimately, diagnosis is confirmed by histopathology and resolution of symptoms with metronidazole. Our case highlights the unusual diagnosis in an immune-competent patient with a puzzling work up.

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